The skinache syndrome

Carl-Fredrik Bassøe MA MD PhD

J R Soc Med 1995;**88**:565–569

Keywords: *skin; chronic pain disorder; diagnosis; treatment*

SUMMARY

Chronic pain of unknown aetiology, and characterized by cutaneous trigger points, has been coined the skinache syndrome. The treatment of the skinache syndrome was evaluated in 94 patients by two independent methods 2 years after treatment. After one subcutaneous injection of lidocaine 68% of the patients were cured. The pain recurred in 27 patients having suffered for an average of 2 years. Surgical removal of the cutaneous trigger points cured 77% of the latter patients. The odds ratio of success of surgical treatment versus all other treatments combined was 101.3. The skinache syndrome requires a precise clinical investigation. Even when the origin of the pain in tendons, muscle and adipose tissue is excluded, the skinache syndrome remains a common, debilitating disorder. In contrast to fibromyalgia, the skinache syndrome has a simple and effective cure.

INTRODUCTION

Chronic pain of unexplained origin and aetiology is a major problem in cardiology[1], geriatric medicine[2], gynaecology[3] and general practice[4]. Chronic pain disorders encompass ill defined muskuloskeletal disorders, 'die Pannikulose'[5], fibromyalgia[6,7], lipomatosis dolorosa[8–10] and the idiopathic pain syndrome[11]. Pain can be triggered by stimulating nociceptors in the skin, joints and muscles[12], but in spite of the comprehensive knowledge of the anatomy, physiology and pharmacology of pain mechanisms, the origin of the pain in chronic pain conditions often remains an enigma[1,12,13].

An effective cure for chronic pain remains to be found[14], and a non-specialist multidisciplinary approach is often the only alternative[15]. Lidocaine injections have been surprisingly effective in the treatment of some pain conditions[16–18]. Unfortunately, the effect is transient in about 20% of the patients, and the source of the pain is unknown. In the present study, stringent criteria are used to identify the dermal source of pain. The skinache syndrome is defined, and the results of surgical treatment are presented.

PATIENTS AND METHODS

Patients

The patients were recruited from a family practice localized in the city of Bergen, Norway. All of them had sought medical treatment because of a pain condition.

The PROMED Institute, Bergen, NORWAY.

Correspondence to: Carl-Fredrik Bassøe, Lyngvegen 14b, N-5082 Eidsvågneset, Bergen, Norway

The skinache syndrome

Trigger points were defined as areas of the body surface measuring about $0.8 \times 1.0\,cm^2$, i.e. the area of a fingertip, from which pain could be elicited or worsened by pressure or pinching of the trigger area[18]. The hyper-sensitivity of the trigger points were corroborated using neighbouring and contralateral cutaneous spots as references.

Care was taken to discriminate cutaneous origin of the pain from a source in the underlying tissue. An area of about 1–2.5 cm in diameter encompassing the trigger point was marked by a pen. The marked spot was moved tangentially relative to the underlying tissue. Pressure was applied to the marked spot and to the site that had been beneath the marked spot. If pain was elicited from the marked spot only, the patient was included in the study.

In order to corroborate the dermal origin of the pain, 1–2 ml 5% lidocaine chloride containing 5μl adrenalin (Astra Farmasøytiske, Skårer, Norway) was injected beneath the trigger area in the superficial part of the subcutis. If analgesia was obtained within 30 s the patients were considered to suffer from the skinache syndrome.

Patients with the skinache syndrome were told not to wash the marked area. If the pain recurred, a new appointment was made within the next 1–7 days. On the following consultation, the patients were blinded to the marked area. If the pain could be evoked from the marked trigger point, the marked area was anaesthetized. If analgesia was obtained after 30 s, the patient was immediately subject to surgery.

Lidocaine injections were used as a routine in the differential diagnosis of pain. The patients were told that the outcome of the treatment with local anaesthesia was unknown. If analgesia was obtained by lidocaine, they

Table 1 **The localization of 107 painful spots in 94 patients suffering from the skinache syndrome. The values were obtained from the computerized medical record, and expressed as a percentage of painful spots in each region of the total number of painful spots**

Region	*Skinache (%)*
Scapula	11.2
Medial to the scapula	2.8
Shoulder	3.7
Elbow	0.9
Wrist	0.9
Finger	0.9
Anterior chest wall	7.5
Abdominal wall	13.1
Lumbar	6.5
Gluteal	8.4
Sacral	3.7
Thigh	6.5
Knee	29.9
Ankle	3.7

were told that the most likely origin of the pain was cutaneous, and that, apart from the suffering, the disorder was innocent. Patients undergoing surgery were informed about the procedure, and told that the outcome was unknown. Verbal, informed consent was obtained from all of the patients.

Surgical procedure

The cutis of the marked area measuring about 1–2.5 cm in diameter and encompassing the trigger point was either removed or surgically disconnected from the underlying subcutaneous tissue. Six lipomas beneath trigger points were investigated, and removed. Specimens were fixed in 4% formalin (v/v), stained by haematoxylin-eosin and examined by light microscopy by two pathologists.

Medical records and data retrieval

A problem-oriented, computerized medical record was used to store the clinical history, investigations, treatments and treatment responses[19]. The patients with the skinache syndrome were recovered as described before[20]. A copy of each patient record was printed and evaluated. In addition, the medical records were subject to automated natural language processing[21].

Questionnaire

A questionnaire was sent arbitrarily to 24 of the 27 patients that had undergone surgery after I had resigned from the practice. The questionnaire contained information stating the anonymity and confidentiality of the information requested. An accompanying letter contained the following statement:

> If the method does not work, no more patients should be treated that way. If it does work, however, the method may be important for the treatment of many other patients. I therefore urge you to answer the questions below as truly and accurately as you can.

All the answers requested were of the form yes/no/don't know.

Statistical methods

The effect of surgical treatment was estimated by the odds ratio[22] (p 318–319). The medical records were compared to the questionnaire using Yates corrected χ^2 [22] (p 313–315).

RESULTS

Ninety-four patients with the skinache syndrome were retrieved from the database. The frequency of the syndrome was 3.1%. The distribution of the trigger points is shown in Table 1. The number of trigger points per patient was 1.6+1.1 (mean+SD) (range 1–5). The age distribution was unimodal with mode 60–69 years, and a female:male ratio of 3.8.

According to the medical records the skinache syndrome had lasted more than 3 months in 61% and less than 3 months in 39% of patients that were cured by lidocaine alone. In the operated patients, the pain had lasted for 88.8 ± 99.3 months (mean $\pm$ SD, range 0.4–20 years).

Effect of lidocaine and surgery

According to the medical records 64 of 96 patients were cured by one lidocaine injection. Three patients were not better, and refused further investigations.

According to both the medical records and the questionnaire 76–79% of the patients were cured by surgery. The pain recurred in one patient. Removal of the scar conferred permanent relief. There were no complications. Four of the five non-responders suffered from another chronic pain in the same region as the trigger point.

Lipomatosis non-dolorosa

Prepatellar lipoid hypertrophy is a cardinal sign of lipomatosis dolorosa. Cure was obtained by surgery in four of six patients without removing the hypertrophic adipose tissue. In six other patients the trigger point was overriding a lipoma. Manipulation of the lipomas during the operation proved them to be indolent. The patients experienced little discomfort during the resection of the unanaesthetized lipomas.

Table 2 **Association between the skinache syndrome and various disorders. The data are from the computerized medical record. The number of patients with the skinache syndrome alone (SSA), or skinache and another disorder (SAD) were automatically obtained. Statistics using the Mantel–Haenszel test are based on comparing each disorder with the controls. The level of statistical significance is shown in the right-most column**

	Men			Women		
	SAD	SSA		SAD	SSA	
Controls	8	309		12	170	
Obesity	4	30	0.01<P<0.025	13	49	P<0.001
Hypertension	6	53	0.01<P<0.025	16	65	P<0.001
Social problems	1	32	NS	9	52	0.01<P<0.025
Anxiety states	1	33	NS	4	53	NS
Dysthymic	0	22	NS	3	36	NS
Sleep disorder	1	25	NS	10	53	P=0.05

NS=No significant association

Treatments before surgery

A total of 71 therapeutic trials of physiotherapy, NSAIDs and/or codeine had been performed in 21 patients who answered the questionnaire. A good response was reported by 8.5% of the patients. About half of the remaining had not benefited from the treatments. The odds ratio of success of surgery versus all the other methods was 101.3. The therapeutic gain of surgery was 84%.

The skinache syndrome and other disorders

Automated language processing was performed on the computerized medical records. The symptoms and signs in cancer, angina, and intra-abdominal conditions were compared to those in the skinache syndrome. Only the words related to trigger points, surgical procedure and lidocaine injections discriminated the skinache syndrome from angina, cancer angina or intra-abdominal conditions.

The occurrence in the same patient of the skinache syndrome and other disorders was studied using the computerized medical record (Table 2). Patients seeking a routine checkup were used as controls. There was a slight excess of adipose- and hypertensive patients among the patients with the skinache syndrome, but associations with psychiatric conditions and sleep disorders were not found.

Histopathology

Histopathological examination was performed on 17 skin specimens. The skin was normal in four. A mild-to-moderate hyperkeratosis and/or a light unspecific inflammatory reaction were observed in 13 specimens. These findings were expected from the first lidocaine injection. Nerve fibres were not seen in the lipomas, but two had nerve fibres in the stalk.

Correlation between medical records and questionnaire

The time of follow-up between the operation and the questionnaire was 19.1 ± 14.8 months (mean $\pm$ SD). The medical records and questionnaire were researched at the same time. Using Yates corrected χ^2 the two sets of data were similar.

DISCUSSION

The present study has delineated a new, common clinical entity characterized by chronic pain originating from trigger points in the skin. About 68% of the patients were cured by one injection of lidocaine, in accordance with the findings of others[16–18]. The effect of lidocaine was transient in 29%. In these patients the dermal nociceptors in the trigger area were surgically disconnected from their afferent nerves[23]. Seventy-seven per cent of the latter patients stayed free from pain. This is a novel finding of considerable clinical importance.

Localizing the tissue source of the pain is not attempted in most chronic pain conditions[1,3,24–26]. In the skinache syndrome the anatomical origin of the trigger points is in accordance with the distribution of cutaneous nociceptors[12,13,27,28]. A diminished threshold or persistent autonomous activity in peripheral nociceptors may explain the pain[29]. Trigger points are also observed in adiposis dolorosa, fibromyalgia, fasciitis and myogenic pain[30], but these sources were ruled out. Clearly, adiposis dolorosa, myogenic pain, fibromyalgia and fasciitis are inappropriate terms for the pain syndrome studied here.

When appropriate treatment is directed at clearly defined physical disease, it is likely to be successful[31]. Acupuncture, intense cold, injections and other mechanical treatments are used against chronic pain[32]. They may

damage peripheral nociceptors, and reduce pain by a mechanism similar to that of lidocaine injection and surgery. With the present treatments, the skinache syndrome has a higher probability of cure than adiposis dolorosa, myogenic pain, fibromyalgia and fasciitis[4,7,9,33], suggesting that the methods were appropriate.

The pain may have been caused by psychological mechanisms. In that case it would be difficult for the blinded patients to localize the trigger points accurately, which they in fact did. Furthermore, there was no correlation between the skinache syndrome, anxiety or dysthymic states. Fibromyalgia may be associated with sleep disorders and depression[11,34], but that was rare in the skinache syndrome. In addition, I minimized expectations, and warned against inappropriate answers to the questionnaire. Finally, analysis of computerized medical records abolishes the confounding Hawthorne effect[35]. Accordingly, it seems unlikely that the pain and effect of treatment were due to psychological mechanisms alone.

The long-term observation before and after treatment strongly suggests that the sudden effect of lidocaine and surgery were not due to the natural history of the disease. The patients did report both transient and lasting effects, ruling out selective reporting. The high odds ratio of surgical effect versus earlier treatments makes regression to the mean unlikely. The two methods used to evaluate the results correlated closely, and the computerized medical record has been shown to contain reliable information[21,36,37]. Taken together, the methods and results indicate that the findings in the present investigation are reliable.

This study is neither randomized nor placebo controlled. The operated patients served as their own controls. The treatments before surgery included physical contact and manipulations, but did not cure the pain. Injections and surgery are associated with a strong placebo effect. However, the magnitude of the lidocaine effect and surgery is 68% and 77%, respectively, which is above that expected from placebo alone. Further randomized trials are needed to confirm the present findings.

I conclude that chronic, severe pain may originate in the skin. The pain seems to be relieved by blocking signals from cutaneous nociceptors. The skinache syndrome is a common disorder that needs and deserves further attention.

REFERENCES

1 Katon W. Chest pain, cardiac disease, and panic disorder. *J Clin Psychiat* 1990;**51**:27–30

2 Ferrell BA, Ferrell BP, Osterweil D. Pain in the nursing home. *J Am Geriat Soc* 1990;**38**:409–14

3 Kames LD, Rapkin AJ, Naliboff BD, Afifi S, Ferrer-Brechner T. Effectiveness of an interdisciplinary pain management program for the treatment of chronic pelvic pain. *Pain* 1990;**41**:41–6

4 Frølund F, Frølund C. Pain in general practice. *Scand J Prim Hlth Care* 1986;**4**:97–100

5 Müller W. Die Pannikulose. *Z Rheumaforsch* 1973;**32**:169–76

6 Rice JR. Fibrositis syndrome. *Med Clin N Amer* 1987;**70**:455–68.

7 Wysenbeek AJ, Mor F, Lurie Y, Weinberger A. Imipramine for the treatment of fibrositis: a therapeutic trial. *Ann Rheum Dis* 1985;**44**:752–3

8 Dercum FX. Three cases of a hitherto unclassified affection resembling in its grosser aspect obesity, but associated with special nervous symptoms-adiposis dolorosa. *Am J Med* 1892;**104**:521–35

9 Iwane T, Maruyama M, Matsuki M, Ito Y, Shimoji K. Management of intractable pain in adiposis dolorosa with intravenous administration of lidocaine. *Anesth Analg* 1976;**55**:257–8

10 Kling DH. Juxtaarticular adiposis dolorosa. *Arch Surg* 1937;**34**:599–630

11 Almay BGL. Clinical characteristics of patients with idiopathic pain syndromes. Depressive symptomatology and patient pain drawings. *Pain* 1987;**29**:335–46

12 Hensel H. Reception-perception. In: R Janzen, WD Keidel, A Herz, C Steichele, JP Payne, RAP Burt, eds. *Pain*. Stuttgart: Georg Thieme Publishers, 1972;53–60

13 Iggo A: The case for "pain" receptors. In: R Janzen, WD Keidel, A Herz, C Steichele, JP Payne, RAP Burt, eds. *Pain*. Stuttgart: Georg Thieme Publishers, 1972:60–67,127

14 Levine J. Pain and analgesia: The outlook for more rational treatment. *Ann Intern Med* 1984.**100**:269–76

15 Loeser JD, Cousins MJ. Contemporary pain management. *Med J Aust* 1990;**153**:208–16

16 Bourne IHJ. Treatment of painful conditions of the abdominal wall with local injections. *Practitioner* 1980;**224**:921–5

17 Petersen P, Kastrup J. Dercum's disease (adiposis dolorosa). Treatment of the severe pain with intravenous lidocaine. *Pain* 1987;**28**:77–80

18 Gallegos NC, Hobsley M. Recognition and treatment of abdominal wall pain. *J R Soc Med* 1989;**82**:343–4

19 Bassøe C-F, Sørli WG. A problem-oriented computerized medical record worked, whereas a chronological did not. In: Hansen R, Solheim BG, O'Moore RR, Roger FH, eds. *Lecture Notes In Medical Informatics*. Berlin: Springer-Verlag, 1988;190–4

20 Bassøe C-F. Skinache: A new, common disorder identified using a computerized, problem-oriented medical record. In: Hansen R, Solheim BG, O'Moore RR, Roger FH, eds. *Lecture Notes In Medical Informatics*. Berlin: Springer-Verlag, 1988;195–9

21 Rasmussen J-E, Bassøe C-F. Semantic analysis of medical records. *Meth Inform Med* 1993;**32**:66–72

22 Rosner B. *Fundamentals Of Biostatistics*. Boston, Mass.: Duxbury Press, 1986

23 Brodal A. *Neurological Anatomy*. New York: Oxford University Press, 1981;47–9

24 McCain GA, Scudds RA. The concept of primary fibromyalgia (fibrositis): clinical value, relation and significance to other chronic muskuloskeletal pain syndromes. *Pain* 1988;**33**:273–87

25 Drewes AM. Primary fibromyalgia/fibromyositis. *Nord Med* 1990;**105**:78–81

26 Keefe FJ, Caldwell DS, *et al.* Osteoarthritis knee pain: a behavioural analysis. *Pain* 1987;**28**:309–21

27 Noordenbos W. Some historical aspects. *Pain* 1987;**29**:141–50

28 Sinclair D. Normal anatomy of sensory nerves and receptors. In: A Harrett, ed. *The Physiology and Pathophysiology Of the Skin*. New York: Academic Press, 1973:348–401

29 McMahon S, Koltzenburg M. The changing role of primary afferent neurones in pain. *Pain* 1990;**43**:269–72

30 Ohrbach R, Gale EN. Pressure pain thresholds, clinical assessment, and differential diagnosis: reliability and validity in patients with myogenic pain. *Pain* 1989;**39**:157–69

31 Waddell G, Bircher M, Finlayson D, Main CJ. Symptoms and signs: physical disease or illness behaviour? *BMJ* 1984;**289**:739–41

32 Melzack R, Stillwell DM, Fox EJ. Trigger points and acupuncture points for pain:correlations and implications. *Pain* 1977;**3**:3–23

33 Eisman J, Swezy RL. Juxta-articular adiposis dolorosa: What is it? *Ann Rheum Dis* 1979;**38**:479–82

34 Ahles TA, Yunus MB, Masi AT. Is chronic pain a variant of depressive disease? The case of primary fibromyalgia syndrome. *Pain* 1987;**29**:105–11

35 Nilsen JF, Bassøe C-F. Contraception in general practice analyzed by a computerized medical record. *Scand J Prim Hlth Care* 1987;**5**:217–20

36 Bassøe C-F. Automatisk analyse av journaltekst. *Tidsskr Nor Laegeforen* 1994;**114**:1974–6

37 Bassøe C-F. Automated diagnoses from clinical narratives: A medical system based on computerized medical records, natural language processing and neural network technology. *Neural Networks* 1995;**8**:313-19

(Accepted 22 February 1995)